

Case Report

Placental site Trophoblastic Tumor with Pulmonary and Brain Metastases

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Abstract:

A rare case of Placental Site Trophoblastic Tumor with multiple metastases was managed with initial chemotherapy (EMA-CO) and radiotherapy followed by surgery with good prognosis.

Key Words: PSTT, EMA-CO

Introduction:

Placental site trophoblastic tumor (PSTT) is a very rare form of gestational trophoblastic disease (GTD), consisting predominantly of intermediate trophoblast and has some unique features such as low serum concentration of β -hCG and high human placental lactogen (hPL), relative insensitivity to chemotherapy and late metastasis.¹

Case Report:

A 25 years old, Para 2+0 with two normal deliveries, having the last child birth 4 years back, was referred from periphery to the outpatient department of G&O in NRS Medical College and Hospital, as a diagnosed case of PSTT. At the time of admission, her chief complaints were irregular vaginal bleeding, cough and headache for last one year. There was no preceding history of amenorrhea.

On examination, she was thin built, severely anaemic with a BP of 100/60 mm of Hg and chest clinically normal. Abdominal examination revealed a firm, irregular, suprapubic mass corresponding to 14-16 weeks of gestation with restricted mobility. Cervix and vagina appeared normal on speculum examination. Bimanual vaginal examination established a bulky uterus of 14-16 weeks size with the os open and moderate vaginal bleeding. A cystic mass about 8x6 cm was felt through the left fornix, adherent with uterus.

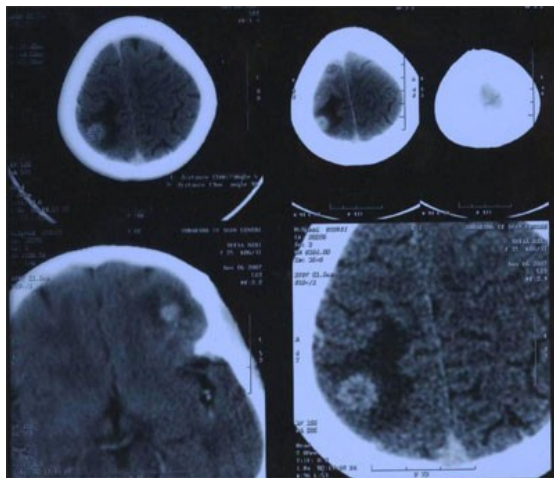


Fig 1: CT Scan showing Brain Metastasis

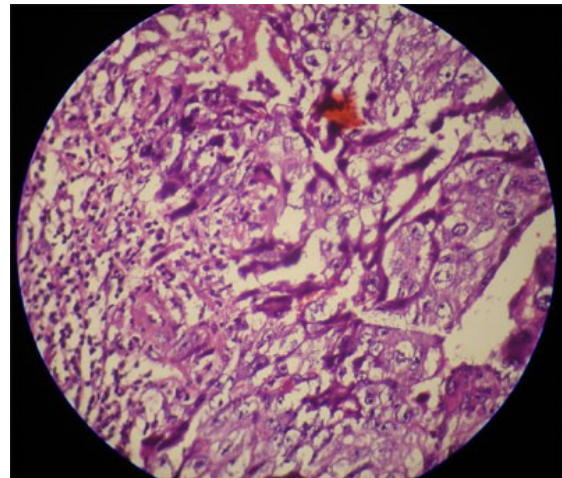


Fig 2: Histopathology of PSTT

Investigations revealed that her haemoglobin was 2.4gm % and LFT, urea and creatinine were within normal limits. Ultrasound examination of the whole abdomen and pelvis showed one heterogeneous mass (8.1x6.4cm) in the anterior and upper part of the uterus, indenting the bladder wall. On color Doppler study the mass was found to be highly vascular. Both ovaries were slightly enlarged with multiple cysts. Chest X-ray showed bilateral multiple opacities. CT scan of the brain revealed two enhancing nodules, one at the left basi-frontal region (7x8mm) and another at right the post parietal region (11x13 mm) with peri-lesional edema and minimal regional mass effect. Endometrial biopsy confirmed PSTT. Her β -hCG level on admission was 9700 mIU/ml while the previous β -hCG was 945mIU/ml. On the basis of clinical, laboratory, radiological and histopathological findings, the case was diagnosed as metastatic PSTT.

After resuscitating her with 6 units of blood transfusion and taking into consideration of her poor general condition and brain metastasis, we started chemotherapy (CT) on her. She received 7 cycles of CT with EMA-CO regime, intra-theal methotrexate (10mg twice a week, 6 doses), cranial irradiation (10 exposures) and local irradiation of abdominal mass (25 exposures) before we opted for surgery. Laparotomy with total abdominal hysterectomy was undertaken. Uterus was found to be bulky and a necrotic solid growth, 5-6 cm in diameter, was found protruding through the anterior uterine wall near left cornu which was adherent to urinary bladder. The mass was relatively avascular and dissection from bladder wall was surprisingly easy with minimal bleeding. However, the histopathology was inconclusive. She was put on four cycles of postoperative chemotherapy. Although her β -hCG level touched baseline after two cycles, two more cycles of CT were given to reduce the chance of recurrence.

Discussion:

Placental site trophoblastic tumor (PSTT) is a very rare and unique form of gestational trophoblastic disease (GTD).¹ There are over just 200 cases reported in literature. PSTT differs histologically and immunologically from gestational choriocarcinoma.¹ Clinical behavior of PSTT varies and prediction of its biological behavior remains difficult. WHO prognostic scoring system also does not correlate well with clinical course of PSTT. However, poor prognostic factors are an interval of >2 years from known antecedent pregnancy, Mitotic Index > 5/10 HPF and extensive necrosis and extension outside the uterus. Increased uterine volume significantly increases the chance of metastasis.² The most common presenting symptoms are irregular vaginal bleeding with or without preceding amenorrhea.³ Diagnosis of PSTT is confirmed by histopathology report. However, persistently low level of β -hCG or elevated hPL along with unexpected resistance to CT raises the suspicion of the disease. It can present with galactorrhea, nephrotic syndrome or just raised serum β -HCG. Metastasis at presentation occurs in 10-15% of patients and recurrence develops in 10% cases. Surgery is the mainstay of treatment in non-metastatic PSTT. Present day advances of CT has a distinct role in the management of PSTT. Radio-therapy is especially effective for metastatic disease.⁴

Conservative therapy by combination CT without hysterectomy may be an alternative for patients desiring future fertility.¹ Bonnazzi et al⁶ reported that one of their patients, treated medically only with EMA-CO, had complete recovery. J-H Nam et al⁵ have reported two cases of PSTT treated successfully by CT followed by curettage without definite surgery. The first line CT regimen is EMA-CO, as reports have shown complete response with this regimen. For EMA-CO refractory cases, second line

CT is EP/EMA. The most recent data from the Charing Cross Hospital, UK and other centers suggest that EMA/EP is the most effective treatment for metastatic or recurrent PSTT. Clinical outcome of PSTT, reported in the literature, are highly variable. All cases of metastasis to vital organs e.g. brain, result in mortality despite all forms of treatment.¹ Our patient, in spite of poor prognostic factors (interval more than 4 years, brain metastasis), tolerated the treatment well with complete remission and she is alive and well till date, one year after completion of treatment. Because of rarity and its successful outcome, this case deserves reporting.

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